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CASE REPORT

Exophiala dermatitidis infection in non-cystic fibrosis bronchiectasis

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KEYWORDS
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Summary A 54-year-old female presented with an exacerbation of right middle lobe bronchiectasis. A bronchoscopic bronchial washing and repeated trials of sputum culture consistently recovered no other infectious agent except *Exophiala dermatitidis*. Her illness was improved by administrations of intravenous miconazole and nebulized amphotericin B when sputum cultures yielded no fungi, demonstrating a pathogenic role of the fungi. The present case illustrates *E. dermatitidis* as a pathogenic agent in non-cystic fibrosis bronchiectasis.

Introduction

Exophiala dermatitidis infection is known to manifest subcutaneous lesions, often in extremities, in immunocompetent persons, whereas more severe infections, such as brain abscess and even systemic infection, can occur in patients in whom immunological defense mechanisms are compromised.[1,2] It is becoming clear that fungi play a significant role as one of respiratory pathogens in patients with cystic fibrosis.[3–5] Here we describe a patient with non-cystic fibrosis bronchiectasis where the fungi were the only organism recovered from a lower airways infection. The infected system was ameliorated by administrations of antifungal agents, demonstrating a similar role in bronchiectasis other than cystic fibrosis.

Case report

A 54-year-old woman presented with increased cough and sputum production. Four years ago she was diagnosed as having right middle lobe bronchiectasis, and had been stable since diagnosis. Her medical history was negative except for bronchiectasis and family history was unremarkable. There were no physical findings. She was unable

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to identify any triggers for her increased cough and sputum production. Laboratory data included WBC; 6900/μl, CRP; 0.16 mg/dl. A thoracic computed tomography (CT) (Fig. 1) revealed right middle lobe bronchiectasis with accompanying infiltrates. Infiltrates were also noted in right S6. Culture of expectorated sputum and a bronchoscopic bronchial washing consistently yielded a growth of yeast-like fungi. No other pathogens including mycobacteria were recovered. Blood β-D-glucan level as 41 (normal: <10) pgml, and galactomannan (latex agglutination) was negative.

Subculture of the fungi on Sabraud agar produced woolly to cottony colonies, colored gray to black, on 30th day at 27 °C (Fig. 2). The isolates did not utilize KNO_3. Conidiogenous cells were intercalary or free and flask-shaped. Annellafed zones were elongated and relatively wide, producing ellipsoidal to sub-cylindrical conidia. From these morphological and biological findings, isolates were identified as *E. dermatitidis*.

Intravenous miconazole (400 mg/day) and inhalation amphotericin B (20 mg/day) was initiated. One month of the treatment resulted in alleviated symptoms and an improvement in radiological findings. Repeated sputum culture did not yield the fungi, suggesting the fungi as the causative agent of her illness.

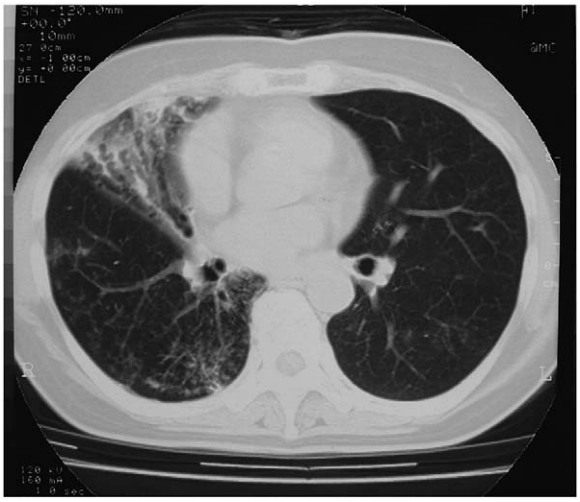

Figure 1 Thoracic CT presenting right middle lobe bronchiectasis accompanied by infiltrates, and irregular infiltrates in right S6.

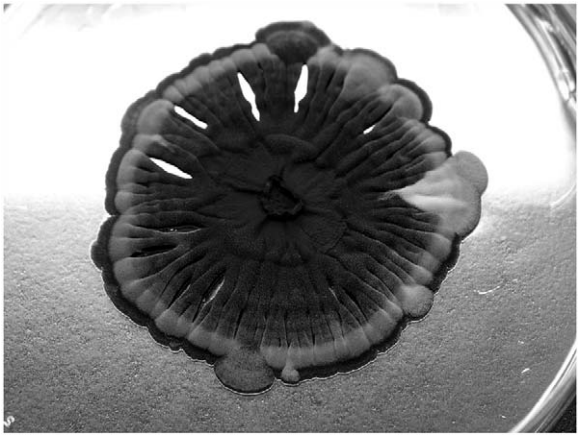

Figure 2 Subculture of the fungi on Sabraud agar producing woolly to cottony colonies, colored grey to black, on 30th day at 27°C.

Discussion

The role of *E. dermatitidis* in the lower airways infection has become increasingly clear, especially in patients with cystic fibrosis. The spectrum of complication for *E. dermatitidis* in cystic fibrosis ranges from colonization[4,6] to invasive infections.[3,5,7] The fungi have also been reported to cause lung disease as a part of systemic infection in patients with compromised immunity.[2] In the woman presented here, *E. dermatitidis* is considered to have caused invasive lung infection because: (1) the organism is the only pathogen repeatedly isolated from airway samples, and (2) clinical and radiological improvement was observed after antifungal treatment when the organism was eradicated.

A prospective study has demonstrated colonization of the fungi in a significant proportion of patients with cystic fibrosis with the application of a specific culture method.[4] Although subculture on Sabraud agar successfully isolated the fungi in the present patient, it is possible that *E. dermatitidis* infection is missed in other clinical settings because of fastidious nature of the fungi in growth. A case where the fungi caused hemoptysis in a patient with non-cystic fibrosis bronchiectasis[7] and the present case imply that there may be many other patients with underlying bronchiectasis in whom *E. dermatitidis* infection are not diagnosed.

Intravenous miconazole in combination with amphotericin B inhalation cleared the symptoms and fungi from the sputum in the present case. Although there can be exceptions,[7] currently available antifungal agents, including amphotericin B, flucytosine, itraconazole, and voriconazole, have yielded clinical improvement in affected individuals.[2,3,5,7] Development of a guideline for optimal diagnosis and treatment should be established because of the potential of substantial numbers of under diagnosed affected individuals.

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